NIH -- W1 J0669R

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ATTN: SUBMITTED: 2001-12-03 15:02:14 2001-12-04 19:09:42 PHONE: 301-496-4563 PRINTED:

REQUEST NO.: NIH-10082252 SENT VIA: LOAN DOC FAX: 301-402-0824 E-MAIL:

5178506

NIH Fiche to Paper Journal

TITLE: JOURNAL OF HAND SURGERY

PUBLISHER/PLACE: Churchill Livingstone Secaucus Nj VOLUME/ISSUE/PAGES: 1996 Mar;21(2):290-2 290-2

DATE: 1996

AUTHOR OF ARTICLE: Amillo S; Schweitzer D; San Julian M

TITLE OF ARTICLE: Monostotic fibrous dysplasia in the hand: a case r

ISSN: 0363-5023

Library reports holding volume or year OTHER NOS/LETTERS:

> 7609631 8683068

SOURCE: PubMed W1 J0669R CALL NUMBER: REQUESTER INFO: AB424

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Monostotic Fibrous Dysplasia in the Hand: A Case Report

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Fibrous dysplasia is a disease of bone in which the involved areas contain immature bone enmeshed in primitive fibrous connective tissue. It is usually monostotic and it is rarely seen in the hand. Schajowicz¹ reported only 3 hand cases in a series of 225 monostotic fibrous dysplasia cases. The Netherlands Committee on Bone tumors² noted only 1 in a series of 235. Milliez and Thomine³ found 5 in 90 cases, while Weaver⁴ and Hayter and Becton⁵ each added 1 case to those reported.

Case Report

We saw a 15-year-old boy with a painless, slowly growing tumor of the hand on the dorsum adjacent to the second metacarpal and limiting index finger flexion. It was a solid tumor, measuring 4 × 3 cm, with deep plane adherence located proximal to the second metacarpal. An x-ray film (Fig. 1) showed deformation of the metacarpal with periosteal reaction. A bone scan was compatible with fibrous dysplasia. A computed tomography scan better defined the extent of cortical involvement and tendon displacement, showing soft tissue involvement. A magnetic resonance image confirmed the presence of soft tissue involvement (Fig. 2). An excisional biopsy was done. Histologic examination showed stromal proliferation of spindle cells without mitoses and fusiform but not atypical nuclei (Fig. 3). There were several foci of reticular bone formation without osteoblastic cell banding. The diagnosis was fibrous



Figure 1. Anteroposterior and lateral x-ray films showing index finger metacarpal deformation, periosteal reaction, radiolucent areas in the bone, and an increased soft tissue density.

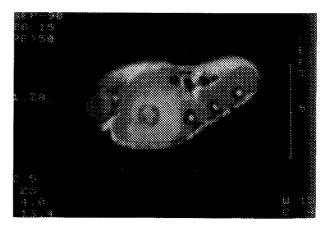


Figure 2. Magnetic resonance image (T1-weighted scan) showing high signal intensity of the cortex with surrounding soft tissue involvement.

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No benefits in any form have been received or will be received from a commercial party related directly or indirectly to the subject of this article. Reprint requests: Professor S. Amillo, Departamento de Ortopedia, Clínica Universitaria de Navarra, Apartado 4209, 31080 Pamplona, Spain.

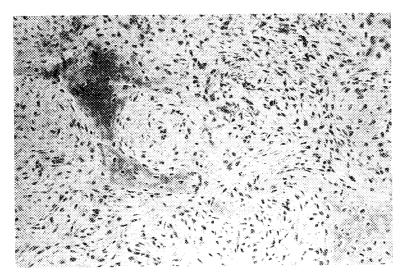


Figure 3. Histopathologic study of the tumor shows stromal proliferation of spindle cells with fusiform nuclei and no mitoses. Several foci with reticular bone formation and without osteoblastic cells were found (original magnification, \times 200; hematoxylin and eosin stain).

dysplasia. The patient returned to his normal activities, but 15 months later had a local recurrence. An angiogram showed a hypervascular lesion without major feeding vessels. An en bloc excision of the tumor with the metacarpal was done, leaving only the distal articular rim. The metacarpal was replaced by an autologous fibula graft held in place by two Kirschner wires.

Four years after surgery the patient shows solid healing and no signs of recurrence (Fig. 4). Although he has only 40° of motion at the metacarpophalangeal joint, he can touch his distal palmar crease and has returned to active sports.

Discussion

Monostotic fibrous dysplasia is rare in the hand skeleton. In this case, the x-ray film appearance suggested a malignancy such as Ewing's sarcoma, but bone scan suggested a benign lesion because the uptake over the metacarpal was uniform and also showed deformation and enlargement. The differential diagnosis was narrowed down to fibrous dysplasia or Paget's disease, and the patient was not of an age for the latter diagnosis.

The great majority of these lesions have been treated by curettage with bone graft. That was not

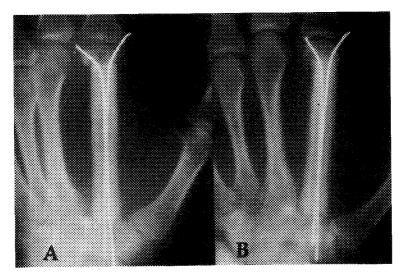


Figure 4. (A) Postoperative x-ray film. (B) Four years after surgery, showing the autograft consolidation.

possible in this case because of the soft tissue component. An index ray resection could have been performed, but we wanted to preserve as much function as possible in such a young patient. The excision with bone graft served to accomplish this end.

References

- 1. Schajowicz F. Fibrous dysplasia. In: Schajowicz F, ed. Tumors and tumorlike lesions of bone and joints. New York: Springer-Verlag, 1981:478–90.
- Netherlands Committee. Fibrous dysplasia. In: Netherlands Committee, ed. Radiological atlas of bone tumours. Paris: Mouton and Co., 1973:479–500.
- 3. Milliez PY, Thomine JM. Rare benign bone tumors and dystrophy in the hand. Review of the literature and report of four cases. Ann Chir Main 1988;7:1125–9.
- 4. Weaver AA. Monostotic fibrous dysplasia: a rare source of hand pain. A report of a case. Nebr Med J 1990;75:43.
- 5. Hayter RG, Becton JL. Fibrous dysplasia of a metacarpal: a case report. J Hand Surg 1984;9A:587.

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